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REVIEW ARTICLE

Neuromuscular scoliosis

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Muscular dystrophy;
Spinal muscular atrophy;
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Summary Scoliosis is a common deformity in many types of neuromuscular disease. Severe spinal curvature can cause difficulty in sitting. Conservative and surgical treatment of neuromuscular scoliosis differs from idiopathic scoliosis, being more complex and with a higher complications rate. Non-surgical measures rarely fully control progressive scoliosis, but aim to prevent spinal deformities secondary to muscular hypotonia or contracture. Twenty-four hour bracing should be adjusted throughout growth, and may induce functional impairment and loss of independence. Corrective surgery requires multidisciplinary management and perioperative screening. Pelvic obliquity is commonly associated with neuromuscular scoliosis, making sitting difficult: correction needs to be considered during surgical planning. The goal of surgical correction is to obtain and maintain a well-balanced spine above a well-positioned pelvis. Preoperative multidisciplinary assessment enables potential problems of terrain to be anticipated. Respiratory function investigation will guide possible non-invasive perioperative ventilation. Nutritional and psychosocial assessment should also be incorporated in this preparation, as should overall postoperative care. Implementing this overall strategic planning can achieve a good surgical and functional result in the vast majority of cases.

Introduction

Spinal deformity is a key issue in the management of several neurological or muscular pathologies. Such pathologies induce control deficits or muscle weakness, which are often the main symptoms. The clinical picture may sometimes be associated with muscle retraction, sensitivity disorder, mental retardation and digestive, cardiac or respiratory problems. Comprehensive management is thus complex but fundamental. The various pathologies have many points in common, in terms both of evolution and of assessment and treatment.

Etiological forms

Table 1 presents the main causes of spinal deviation.

The Scoliosis Research Society (SRS) classifies them as neuropathic, with central or peripheral motor neuron involvement or both, or myopathic [1], a heterogeneous class of pathologies of very variable functional impact [2—5].

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Table 1 Main neuromuscular etiologies of spinal deformity.

Central neurological causes Central motor neuron involvement	Cerebral palsy Hereditary ataxia (Friedreich, etc.) Syringomyelia Other central causes (encephalopathy, Rett's syndrome, etc.)
Peripheral neurological causes Peripheral motor neuron involvement	Acute anterior poliomyelitis Infantile spinal amyotrophy Hereditary motor and sensory neuropathy Hereditary sensory and vegetative neuropathy (familial dysautonomia)
Mixed central and peripheral neurological causes	Medullary lesion Myelodysplasia Myelomeningocele
Neuromuscular junction (motor end-plate)	Myasthenia
Muscular causes	Duchenne myopathy Other muscular dystrophy Arthrogryposis

Such ''secondary'' spinal deformity occurs much more frequently during the course of these pathologies than does ''idiopathic'' scoliosis in the general population: prevalence ranges from 25 to 100% according to etiology (Table 2).

Spinal deformity pathogenesis

Spinal deformity cannot be attributed to trunk muscle weakness alone; indeed, in certain pathologies, trunk muscle hypertonia rather than paralysis is primarily implicated. In central neurologic pathology, for example, spinal deformity may be induced by disharmonious control of trunk musculature around the spinal axis, progressively worsening due to a lack of effective muscular compensation mechanisms (Fig. 1).

Some such deformities misleadingly resemble idiopathic scoliosis, while others present as a long and regular thoracolumbar curve. A long thoracolumbar curve all the way down to the pelvis, inducing pelvic obliquity, is a classic form of neuromuscular spinal deformity (Fig. 2) [7]. In some cases, pelvic obliquity may result from asymmetric retraction of the muscles connecting trunk and pelvis; this is known as pelvic obliquity of ''upper origin'' (Fig. 3A). In other cases, hip posture asymmetry causes retraction, usually in adduction, flexion and internal rotation, predominating on one hip; this asymmetry of retraction induces pelvic malpositioning in the sitting and lying postures, which in turn accentuates the underlying scoliosis: this is known as pelvic obliquity of ''lower origin'' (Fig. 3B). This pathogenic cascade makes it especially important to ensure an optimally symmetrical hip posture, especially in non-walking patients. In some cases, however, retraction co-exists at both spinal and pelvic levels. Finally, kyphotic deformity with trunk collapse is classic in neuromuscular pathology (Fig. 4).

Table 2 General population prevalence of scoliosis according to etiology [6].

Etiology	Prevalence (%)
Cerebral palsy	25
Myelodysplasia	60
Spinal amyotrophy	67
Friedreich's ataxia	80
Duchenne myopathy	90
Medullary lesion (< 10 years of age)	100

Patient assessment in neuromuscular spinal deformity

Clinical assessment

Clinical assessment includes any hyper- or hypotonia in various muscle groups, and joint stiffness. Hip range of motion evaluation should take account of pelvic positioning, which needs to be spatially correct before taking angle measurements. Flexion contracture of the hip can be difficult to examine. The child should be lying flat, prone at the end of the examination table (Fig. 5). Asymmetric hip stiffness may the prime symptom, with a classic rightward or leftward ''drift'' aspect (Fig. 6). This asymmetry causes imbalance in the seated posture. Asymmetric pressure on the ischia creates scabbed areas of hyperpressure (Fig. 7). It also underlies the pelvic obliquity of lower origin, which aggravates the underlying spinal deformity. It also complicates pre- and postoperative positioning of the patient (especially for orthopedic treatment); first-line surgery to correct hip positioning may be needed before treating the trunk deformity itself.

The patient's walking capacity should be assessed [8]. Simply distinguishing ''walking'' from ''not walking'' is not enough. The difficulty lies in assessing patients whose autonomy is limited: indoor walking only, sometimes requiring aids (orthoses, walking frame...) or sometimes just transfer, with a greater or lesser degree of help from a walking-aid or another person. If surgery is liable to endanger walking autonomy, it may be wise to put things off for some months or even years, especially in the case of evolutive

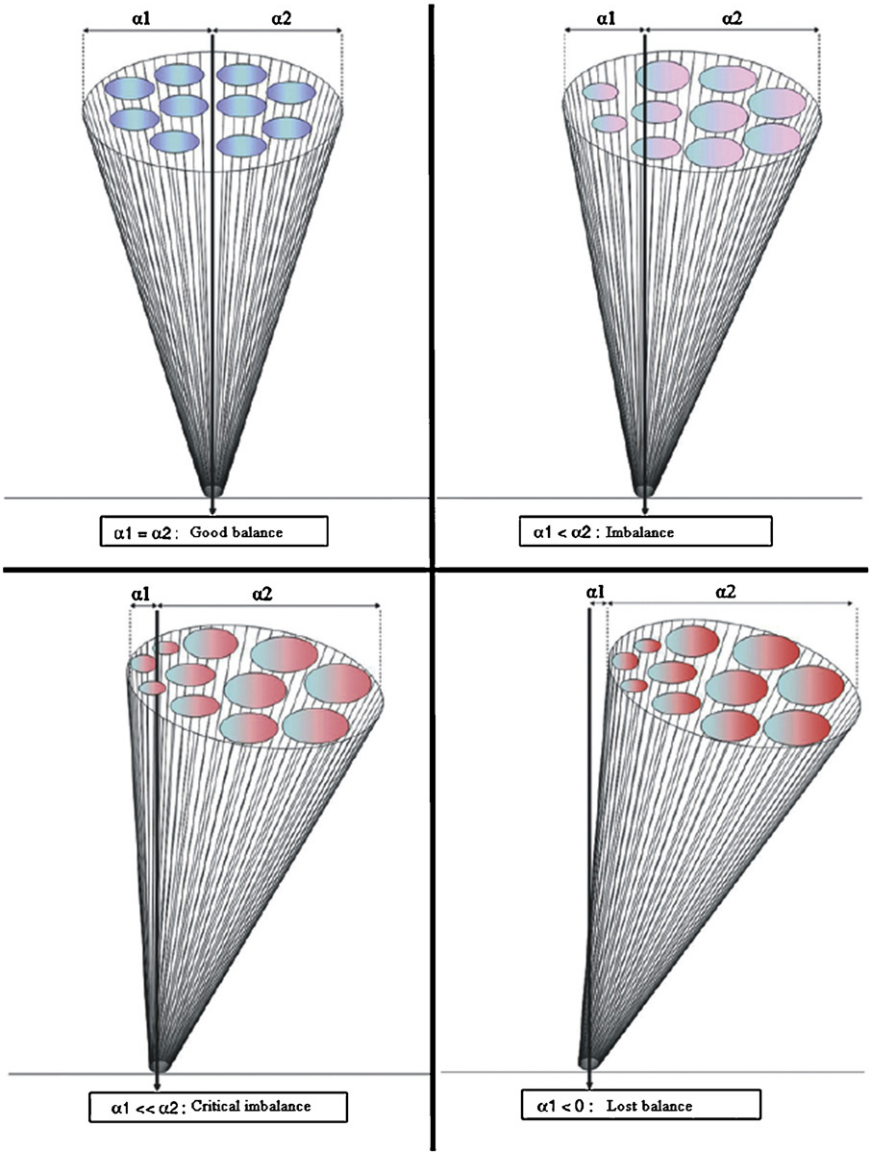

Figure 1 Schema (J. Dubousset/R. Vialle) of progressive trunk imbalance induced by asymmetric application of muscular maintenance forces. Imbalance progresses to the point of inevitable trunk collapse.

neuromuscular pathology, so that vertebral arthrodesis no longer entails a further loss of independence.

Static examination may be performed with the patient sitting at the edge of a table, with the help of another person to maintain the seated posture if necessary. This examination provides better assessment of trunk deformity and of the sagittal and frontal components of the collapse.

Dynamic trunk examination assesses deformity reducibility. The spine should be studied level by level in lateral inclination and in rotational movement. Curvature reducibility can also be judged by trying to raise the patient by the head (Fig. 8).

Prone examination should be systematic, with the patient lying at the end of the table with lower limbs in flexion. This displays residual curvature after eliminating abnormality due to limb-length discrepancy, pelvic asymmetry and gravitational effects. Curvature reducibility can be judged again in lateral inclination, as can the flexibility of the iliolumbar angles.

Respiratory assessment and management

Careful respiratory assessment should be conducted systematically in neuromuscular spinal deformity, for three reasons:

- spinal deformity can impact ventilation mechanics, especially when severe and associated with thoracic hypokyphosis, thoracolumbar kyphosis or rib-cage deformity;
- the underlying neurologic pathology may, in itself or due to its evolution, impair ventilation;

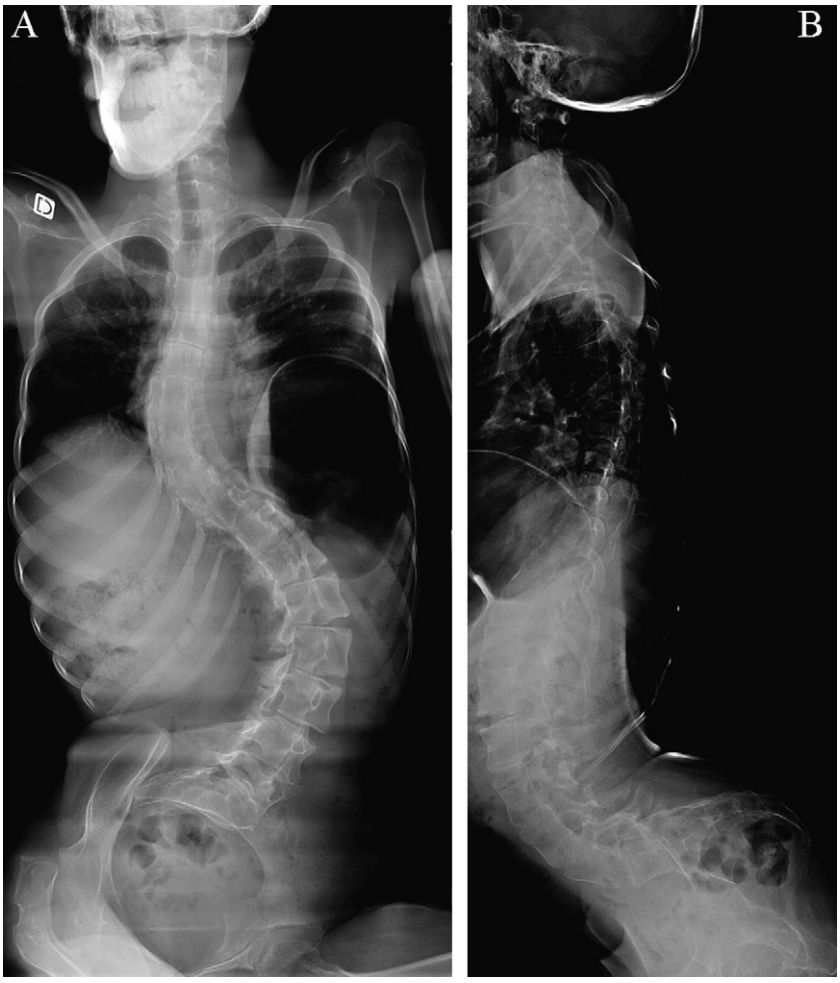

Figure 2 Thoracolumbar scoliosis with pelvic obliquity is a frequent form of neuromuscular spinal deformity.

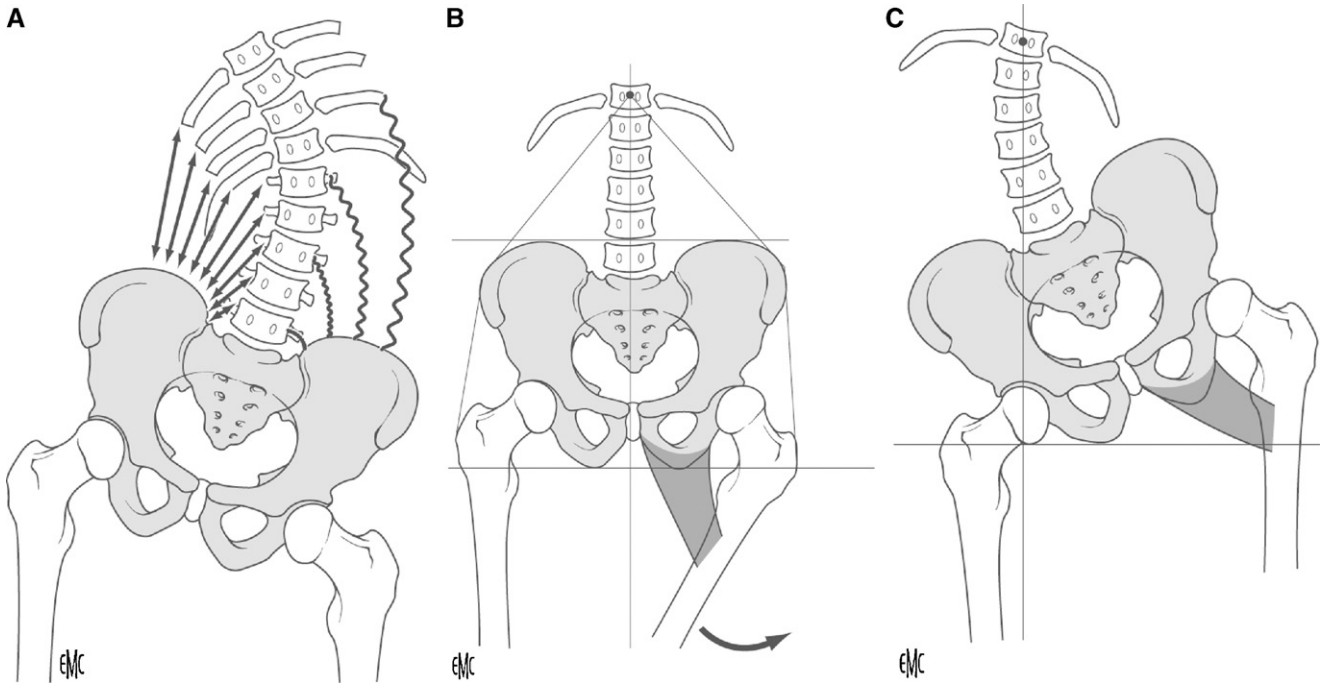

Figure 3 Schema of pelvic obliquity with underlying scoliosis of upper origin (A) and lower origin (B and C).

Figure 4 Evolution of postural hypotonia leading progressively to lumbar kyphosis in a boy with Duchenne muscular dystrophy. A. Lateral spinal X-ray, standing, age 8 years. B. Lateral spinal X-ray, seated, age 10 years.

- treatment, whether conservative or surgical, may have immediate respiratory impact that can sometimes be enduring or definitive.

Respiratory assessment can now rely on objective examinations. Conservative as well as surgical treatment may induce sudden decompensation of a respiratory state that is usually fragile. Associated swallowing disorder, a poorly controlled epileptic state and fragile nutritional status are aggravating factors.

Regular respiratory rehabilitation can be associated to a variety of instrumental techniques, each of which has its own particular benefit (Fig. 9).

Intermittent positive pressure breathing devices (also known as ''pressure relaxers'') such as the Alpha 200 help maintain rib-cage flexibility and improve thoracic ampliation. This technique is intended for stiff spinal deformity with thoracic hypokyphosis or severe chest deformity.

''Cough-assist'' devices are useful in elevated risk of tracheobronchial congestion [9]: hypotonic patients (spinal amyotrophy, muscular dystrophy). They are especially effective when the patient is bed-ridden or fatigued, as during the immediate postoperative course.

Non-invasive ventilation improves spontaneous ventilation quality by a mechanical ventilation aid via an oral or nasal mask connected up to an assisted breathing apparatus [10]. It may be used ahead of heavy surgery and during the first postoperative months in the most fragile patients. Preoperative assessment and family education allow implementation under good conditions [11].

Invasive ventilation via tracheotomy may be required by the severity of respiratory insufficiency or swallowing disorder with recurrent congestion. In patients at high risk of respiratory decompensation, tracheotomy should be discussed among the multidisciplinary team before deciding on any vertebral arthrodesis. This avoids emergency tracheotomy, performed under poor conditions, in case of postoperative respiratory distress.

Cardiac assessment

Minimal cardiac assessment ahead of arthrodesis is mandatory in neuromuscular spinal deformity [12].

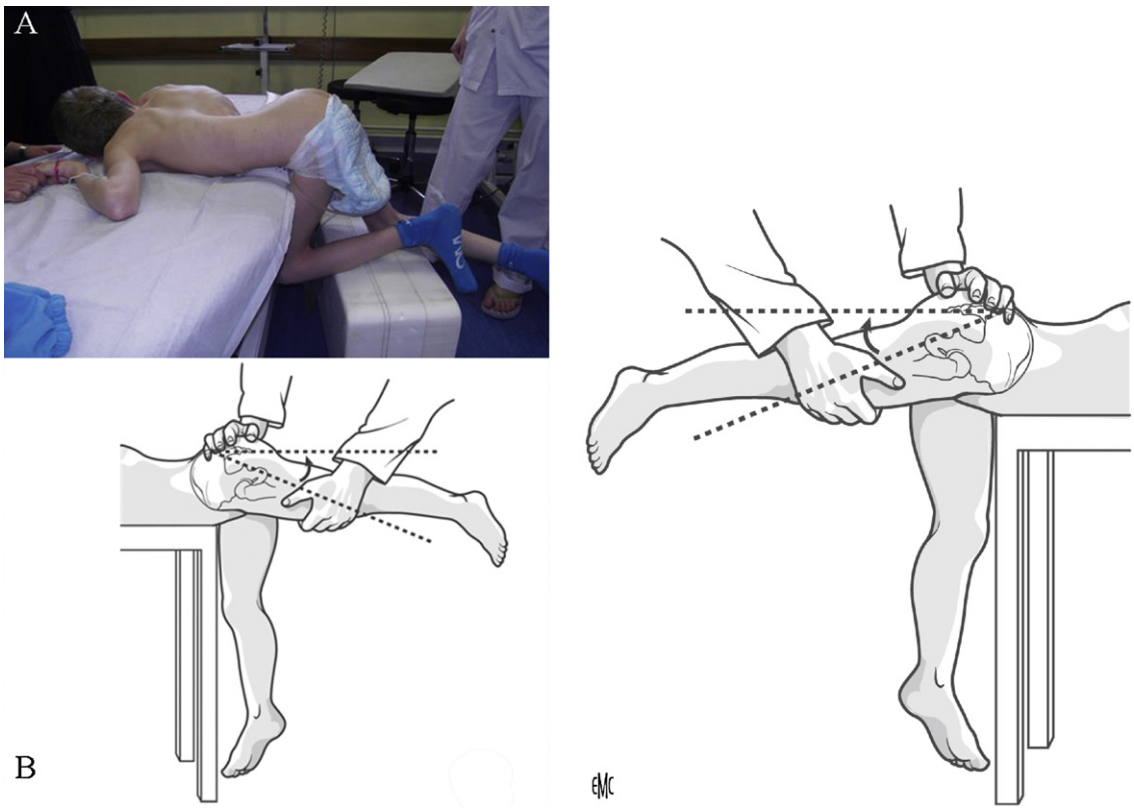

Figure 5 Clinical hip examination with flexion contracture measurement in ventral decubitus with pelvis at table edge. A. Clinical photograph of child's position. B. Diagram of flexion contracture measurement.

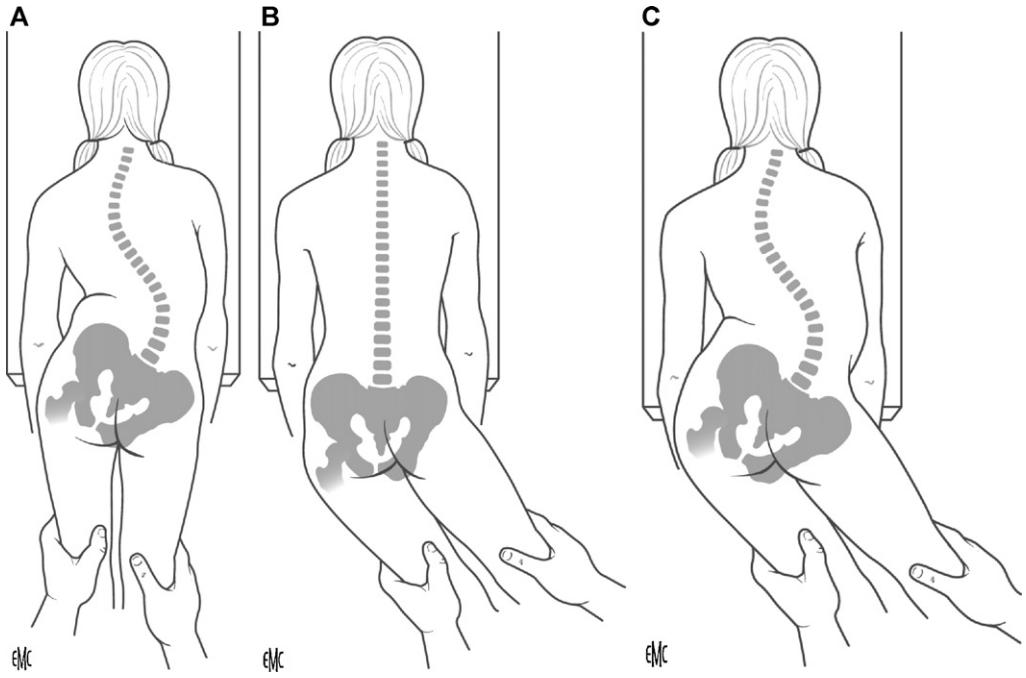

Figure 6 Diagram of pelvic obliquity reducibility assessment in ventral decubitus (superior view). A. Spontaneous position, with pelvic obliquity and hip malpositioning. B. Good pelvic obliquity reducibility. C. Incomplete pelvic obliquity reducibility under asymmetric left lower limb traction.

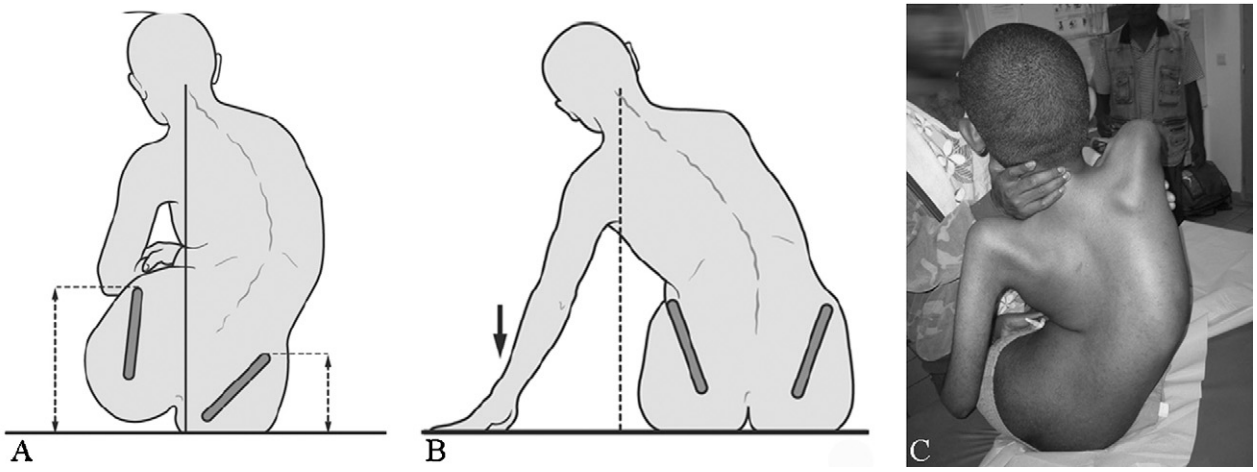

Figure 7 Pelvic obliquity inducing asymmetric stress to ischial weight-bearing points in seated posture. A. Imbalance with excess right ischium pressure. B. Compensatory trunk imbalance by upper limbs. C. Upper limb compensation impossible, requiring help from another person to maintain upright trunk.

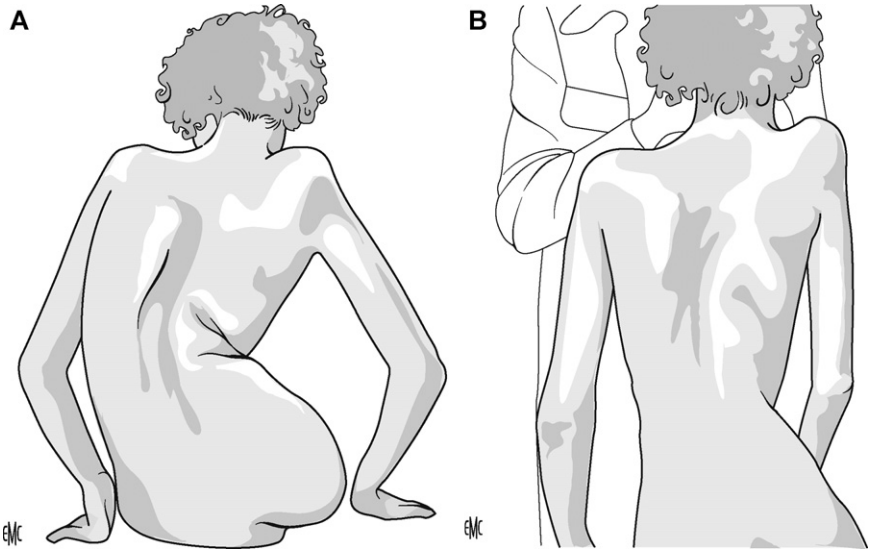

Figure 8 Clinical reducibility of collapse can be explored by manual trunk elongation in seated posture.

Table 3 presents the various types of myopathy with cardiac involvement.

Myocardial contractility is systematically impaired in Duchenne muscular dystrophy. Onset may be sudden, at 10 or 11 years of age; surgical stabilization of the spine should be rapidly undertaken [13,14].

In Steinert myotonic dystrophy, conduction disorder can be screened by Holter or preoperative intracavitary

Table 3 Cardiac involvements in muscular dystrophy.

Usual name	Genetic abnormality	Location	Type of cardiac involvement
Duchenne muscular dystrophy	*DMD* gene	Xp2.1	Cardiac insufficiency
Becker muscular dystrophy	*DMD* gene	XP2.1	Cardiac insufficiency
Emery-Dreifuss muscular dystrophy	*EMD* and *LMNA* genes	Chromosome X, Chromosome 1	Conduction disorder, arrhythmia
Limb-girdle or Erb muscular dystrophy	Polygenic, recessive or dominant	Linked to X	Cardiac insufficiency
Steinert myotonia	*DMPK* gene	Chromosome 9	Conduction disorder
Rett's syndrome	*MECP2* gene	Chromosome X	Cardiac dysautonomia, rhythm disorder

Figure 9 A. Alpha 200 device for positive pressure passive ampliation. B. Cough-assist device for bronchopulmonary decongestion. C. Non-invasive ventilation by nasal mask. D. Invasive ventilation by tracheotomy.

recording. A preoperative electrosystolic training probe may sometimes be necessary to prevent peroperative arrhythmia [15].

Trophic assessment and digestive or urinary disorder

In general, in case of weight-loss or stagnation during the growth period, nutritional deficiency should be primarily suspected, but also impaired respiratory function or depression syndrome. During the months preceding spinal surgery, particular attention should be paid to nutritional management. In the most difficult cases, with history of malnutrition and limited patient cooperation, nocturnal feeding by nasogastric tube or by gastrostomy should be considered several weeks or months ahead of scheduled arthrodesis.

Preoperative management is mandatory for any chronic urinary infection. Urine sterilization and rigorous implementation of a precise catheterization or self-catheterization protocol can limit infection risk, notably in case of medullary involvement (paraplegia, myelomeningocele). In self-catheterization, precise assessment of implementation should precede arthrodesis: altered trunk shape and size may cause difficulty for some patients, altering the technical conditions of self-catheterization.

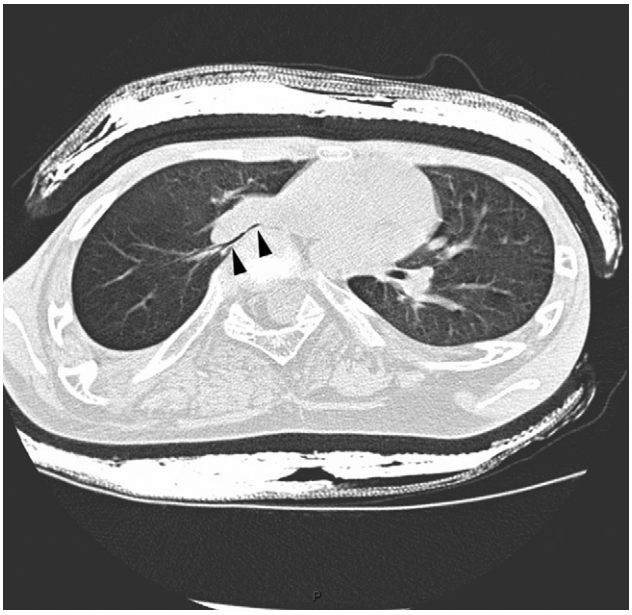

Figure 10 CT slice confirming right middle lobe bronchus stretching (arrows) in contact with vertebral body in girl with evolutive scoliosis with thoracic lordosis.

Imaging

In non-walking hypotonic patients, imaging in the seated posture, where the deformity is accentuated by weight, can be useful. Although realistic with respect to the postural deformity, such imaging is poorly reproductible for diachronic comparison, and images taken in lying posture are preferable.

Specific ''bending'' views are needed to assess stiffening of the different spinal levels. AP views under asymmetric traction (applied to a single lower limb) can assess the frontal reducibility of pelvic obliquity. Complete correction is demonstrated by alignment of the line through the foot of the two sacroiliac interlines with respect to the shoulder line [16].

Preoperative MRI is essential in suspected medullary pathology, even when longstanding (paraplegia, traumatic or non-traumatic quadriplegia), to detect a possible associated syringomyelic cavity that may induce per- or postoperative neurologic aggravation, especially frequent when medullary function is partially conserved [17,18].

Thoracic CT is recommended in spinal deformity with thoracic hypokyphosis or lordosis; the caliber of bronchi anterior to the spinal convexity is often reduced [19]. In severe cases, such bronchial ''stretching'' (Fig. 10) may induce reversible or irreversible atelectasis and reduced lung volume.

Treatment

Avoiding spinal deformity: prevention

Prevention is the keystone of early management of many children and adolescents with neuromuscular disease. Preventing retraction and pathologic posture of the trunk and above all of the limbs is the foundation of comprehensive orthopedic management.

Countering asymmetric hip posture is the most effective way of countering the development of pelvic obliquity and thus of scoliosis of lower origin. Countering flexion contracture of the hips prevents the lumbar and lumbar-sacral region stiffening into hyperlordosis [20,21]. Conversely, progressive retraction of the hamstring muscles induces not only flexion contracture of the knee and increasing hindrance of the upright stance, but also progressive retroversion of the pelvic pedestal and progressive kyphosis of the lumbar spine (Fig. 11).

Evolutive spinal deformity: the role of conservative treatment

In certain pathologies inducing severe muscular deficiency (quadriplegia, type 1 and 2 spinal amyotrophy), orthopedic treatment should be very early. It begins with passive bracing, correcting the spine by traction exerted by a Garchois-type corset between the two fixed points constituted by the pelvis and the skull (Fig. 12).

Some patients with central (cerebral palsy, cerebellar ataxia) or peripheral neurologic disorder (neuropathy) or muscular pathology involving only mild deficiency may be treated using the more conventional Chenau corset or a molded corset for nocturnal hypercorrection. Other neurologic disorders, such as dystonia, are not amenable to treatment by corset.

In most cases, conservative treatment of neuromuscular spinal deformity is only an interim measure awaiting vertebral arthrodesis [22]; it is nevertheless effective in limiting such consequences as impaired pulmonary development and function, enabling future surgery to be as simple and limited as possible.

Evolved spinal deformity: the role of surgical treatment

To operate or not to operate? — Technical and ethical limits

Improvement in surgical and, first and foremost, intensive care, pneumologic, cardiologic and anesthesiologic techniques now allows surgical solutions of varying complexity to be offered to a very large number of patients, including the most fragile. Treatment options should be discussed as early as possible with the patient and his or her family and close environment [23].

Several studies have demonstrated objective postoperative functional improvement based on self-administered quality-of-life questionnaires, confirming the benefit of surgical management, even in the most difficult cases [24].

When to operate?

Some deformities are strongly evolutive and difficult to contain conservatively. Relatively early surgery may thus be justified. Even so, conservative treatment should be continued to the very end if development of the chest and

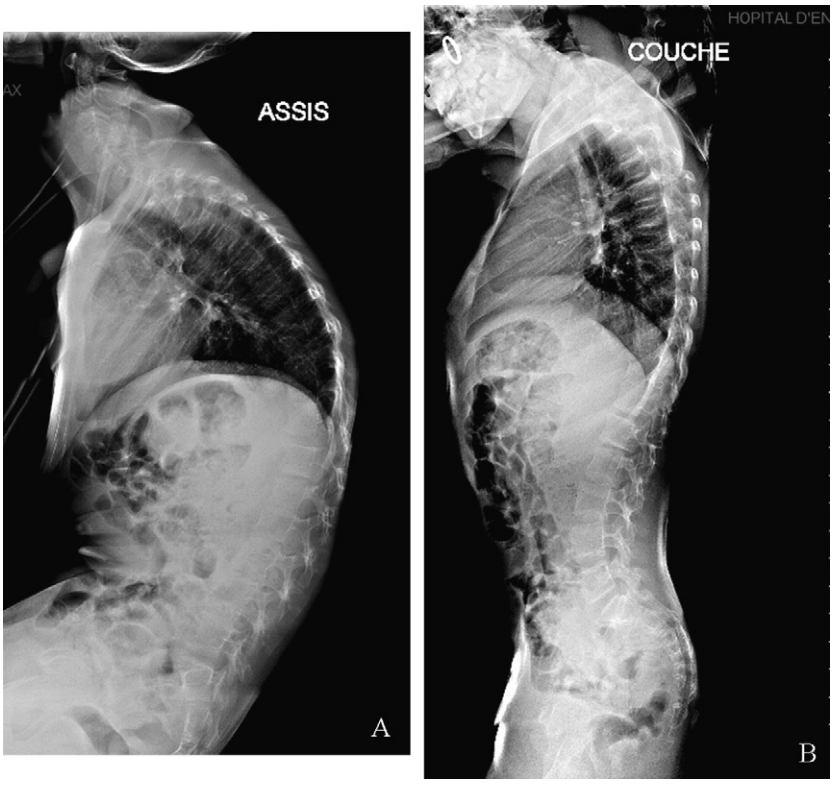

Figure 11 Hamstring retraction inducing pelvic retroversion and lumbar kyphosis in seated posture (A). View in lateral decubitus, hips in extension, confirms complete reducibility and the purely muscular postural origin of the deformity.

increased respiratory capacity can still be obtained by the growth of the trunk. In some poorly controlled deformities, progressive spinal distraction rods, usually associated to classical corset treatment, may be useful. The mechanical resistance thresholds of these devices, however, frequently lead to complications, particularly at the spinal fixation sites.

The clinical and radiological criteria of spinal maturity are a matter of debate in neuromuscular disease [25]. Triradiate cartilage closure is a good sign of axial skeletal

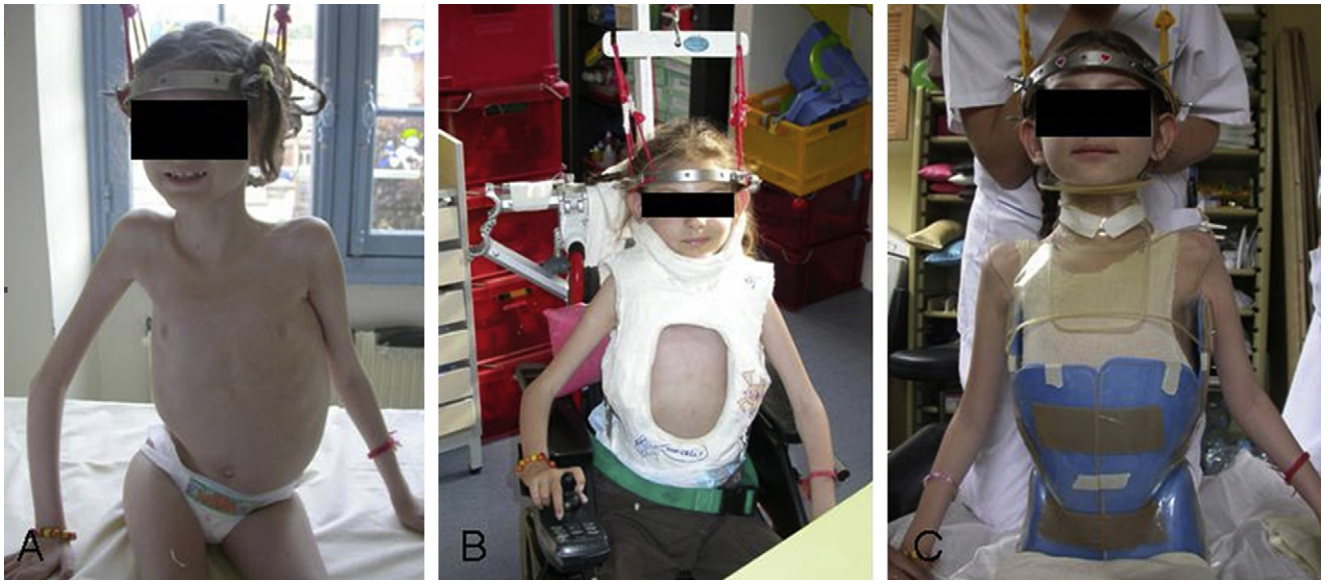

Figure 12 Conservative treatment sequence for severe spinal deformity with pelvic obliquity in a 9-year-old girl with spinal amyotrophy. A. Correction begins with progressive axial traction by cranial halo. B. Supplementary correction of pelvic obliquity and trunk collapse by detraction cast. C. Stabilization of correction in Garchois corset with occipitomental extension.

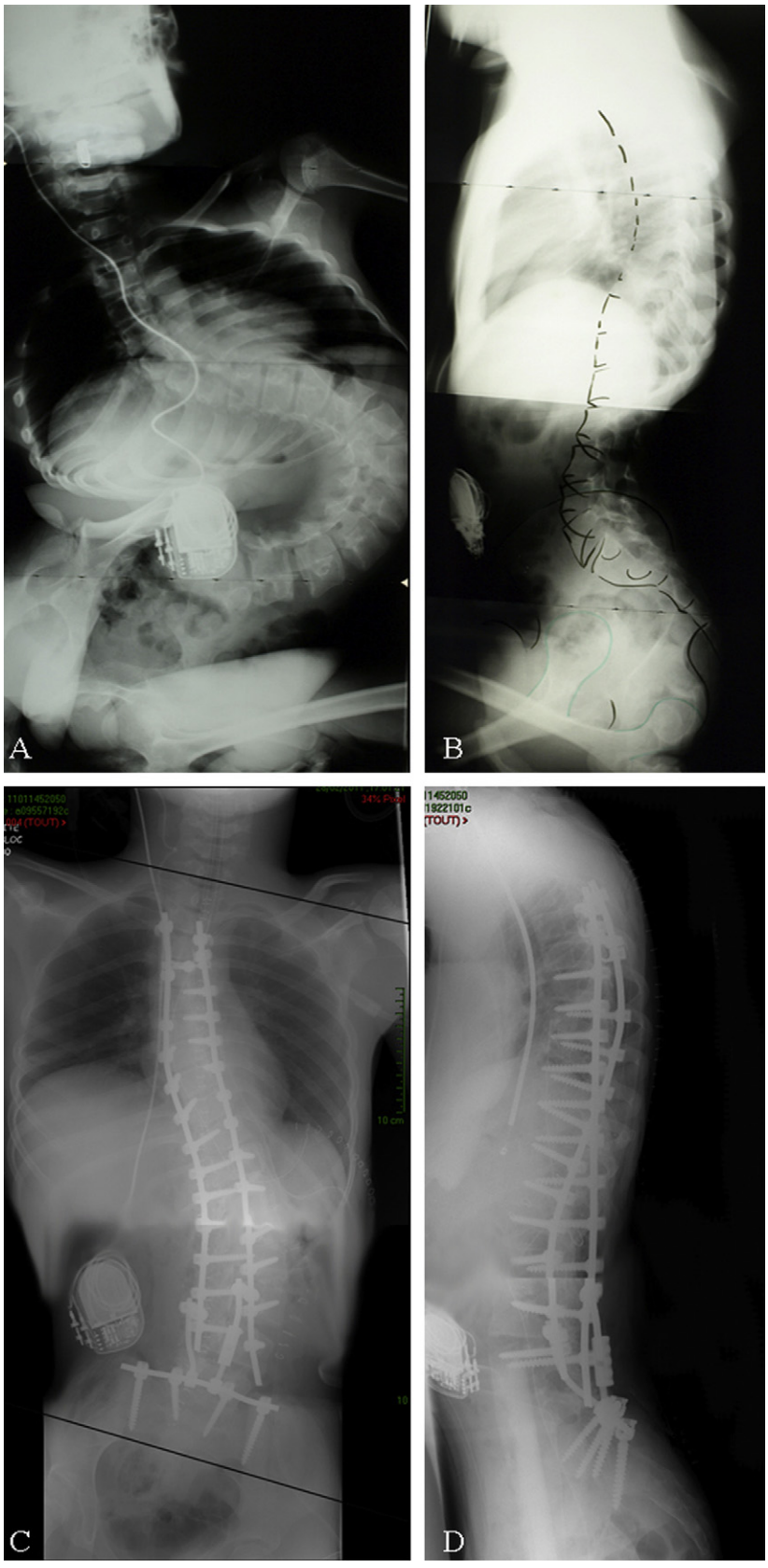

Figure 13 Severely evolved spinal deformity in a 15-year-old boy with cerebral palsy and spastic quadriplegia. A and B. Preoperative X-rays. C and D. Postoperative X-rays after anterior release of lumbar scoliosis convexity and posterior osteosynthesis. Despite initial severity, overall trunk, shoulder line and seated pelvis alignment is satisfactory.

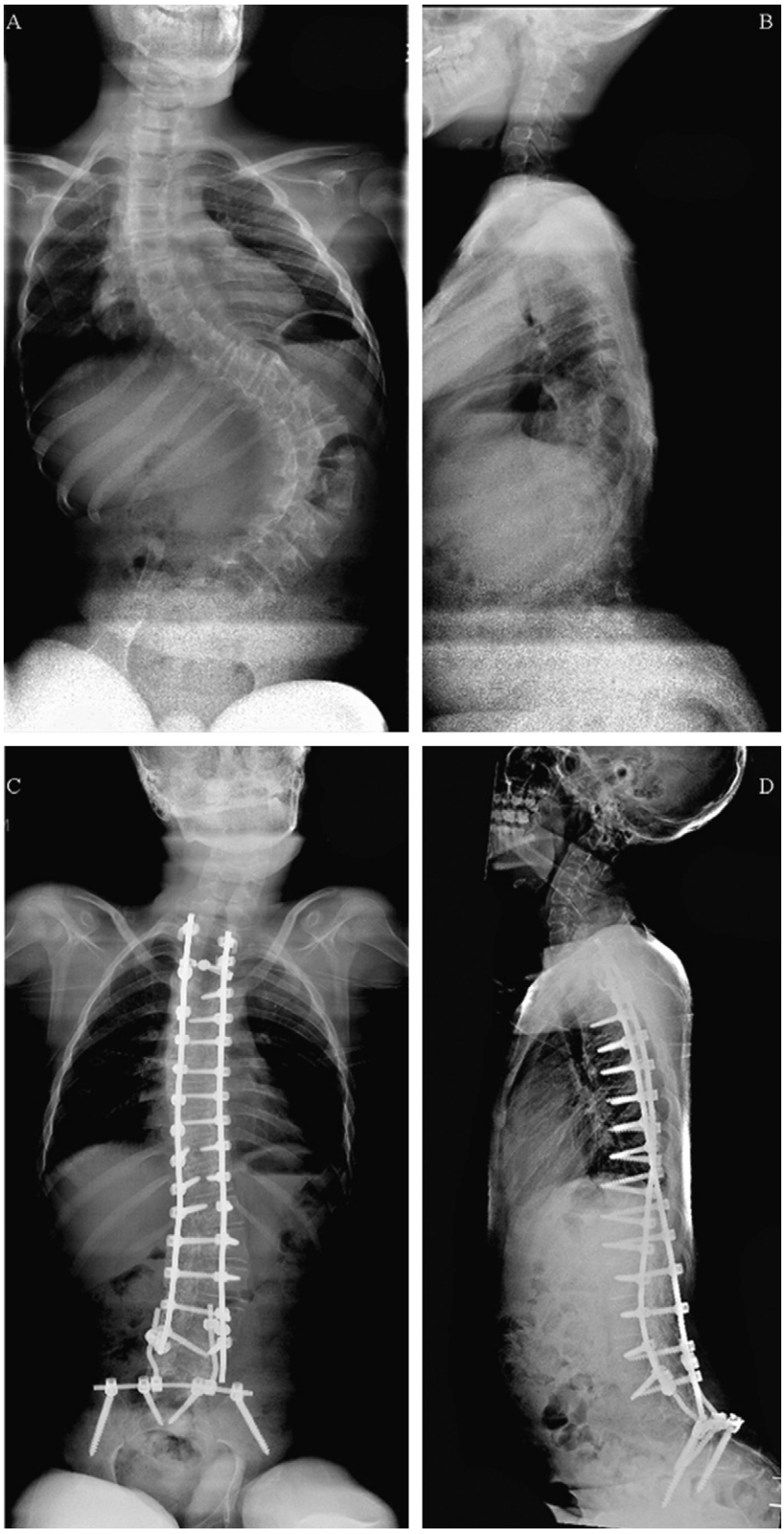

Figure 14 Example of correction by progressive bending and segmental instrumentation by pedicle screws in a 13-year-old boy with congenital muscular dystrophy. A and B. Preoperative X-rays. C and D. Postoperative X-rays.

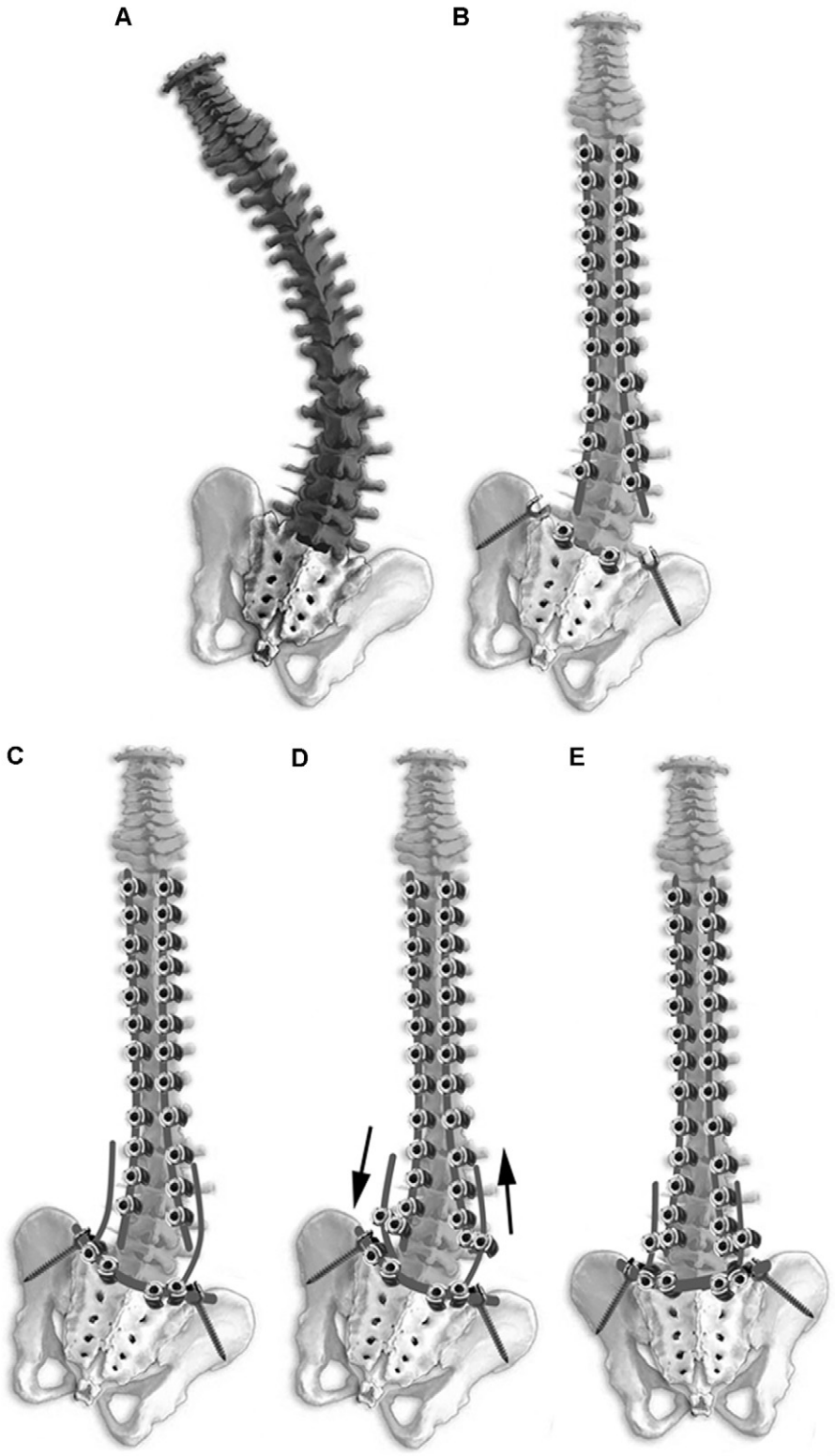

Figure 15 Surgical correction strategy in neurological scoliosis with pelvic obliquity by segmental T-assembly. A and B. Correction of spinal deformity. C. Pelvic instrumentation with 21 pedicle screws in sacrum and two in the iliac wings, connected by a horizontal rod. D. Correction of pelvic obliquity by union connectors between pelvic and spinal assemblies. E. Final result after implant locking.

maturity, but may be late in case of resistant hip dislocation secondary to the neuromuscular pathology.

Optimal scheduling of surgery is thus determined by compromise: not too late, so as to have the least severe and most reducible deformity possible (Fig. 13), but not too early, to limit the risk of thoracic hypertrophy and restricted lung volume.

Specificities of surgical strategy and technique

Surgical techniques. Pediatric spinal deformity surgery has benefited in recent years from technological progress and ever more radical operative strategies developed by teams that also treat adult deformity [26,27]. Segmental pedicular screwing, especially in the apical region, provides 3D spine control [28—31], preventing evolutive deformity following arthrodesis when the operated spine still conserves growth potential (crankshaft phenomenon [32]). It thus avoids preliminary epiphysiodesis. Multiple anchorage with implants at each level of vertebral arthrodesis is a good solution to poor bone quality, with risk of mechanical assembly failure, in osteoporotic patients. Correction byin-situ progressive contouring of the rods is effective, distributing stress over all the implanted levels (Fig. 14). Sublaminar implants such as the Universal Clamp may be applied in the deformity concavity to limit the risk of screw detachment during translation of the concavity toward the stem [33].

The two objectives are optimal correction of the spinal deformity and pelvic obliquity. The aim is to achieve frontal alignment of the pelvic and scapular belts. In severe pelvic obliquity, an effective technique is to position the patient in asymmetric traction on a Cotrel table [6]. Preliminary release of the deformity convexity is justified only in those rare cases where residual pelvic obliquity exceeds 10° on preoperative traction views [16,34]: if the obliquity is reducible on asymmetric traction view, the benefit of preliminary release is greatly outweighed by the risks incurred, notably in terms of postoperative morbidity [35].

Pelvic obliquity correction. Correcting the pelvic obliquity requires the spinal assembly to be extended down to the pelvis. Numerous surgical techniques have been described, and complete mastery comes only after a long learning curve [36].

The pelvic-spinal assembly should enable isolated sequential correction of pelvic positioning with respect to the spinal assembly (Fig. 15). The iliosacral screwing technique developed at the Saint-Vincent-de-Paul Hospital is limited by the need for specific connectors [16,37]. Moreover, poor sacrum bone quality may also greatly impair fixation with such implants. The traditional pelvic extension techniques (e.g. Galvestone) also fail in some cases due to poor anchorage of the spinal assembly down to the pelvis [38,39].

Segmental techniques using pedicle screws or specific iliac extension screws provide good quality anchorage and freedom in the means of fixation. Ideally, several pelvic anchorages (sacral and iliac) are combined and by means of rod segments, so as to ''share'' mechanical risk during correction maneuvers [34,40] (Fig. 13).

Postoperative care. Postoperative management usually begins with a few days or weeks of intensive care; this is a critical period in which postoperative respiratory and infectious complications are not rare. A bivalve protection corset may be useful for early verticalization without stress to the assembly or bone, which remain fragile. Several weeks spent in a rehabilitation center are usually required before discharge home.

Complications

The morbidity induced by surgical correction of neuromuscular spinal deformity is considerable, and far greater than in idiopathic deformity. The SRS database confirmed the high prevalence (> 17%) of general and infectious complications, with a non-negligible mortality risk [41].

Prevention of respiratory complications is primarily based on good respiratory assessment and management well ahead of surgery [42]. Non-invasive ventilation or tracheotomy often succeed in avoiding what could be insurmountable postoperative problems.

Prevention of neurological complications (always possible in these cases) justifies the use of peroperative electrophysiological monitoring, and is technically feasible, especially in case of peripheral neurologic or muscular pathology, but more difficult in central pathologies (cerebral palsy).

Prevention of infectious complications is trickier [43]. Certain risk factors are known and can be attacked (cutaneous colonization, chronic urinary or pulmonary infection, malnutrition, poor oral-dental or cutaneous status, etc.). Other risk factors can be taken into account and dealt with, such as surgery time and peroperative bleeding. Evolution after early surgical revision and prolonged antibiotherapy is favorable in most cases [44].

Severe respiratory or hemodynamic complications leading to death were reported in 0.3% of cases [41], and concerned patients with particularly fragile health status; they should be clearly explained to the patient's family before operating.

Conclusions

Neuromuscular spinal deformity constitutes a broad, heterogeneous nosological category in which the specificity of each case is essentially defined by the repercussions of the underlying neurologic or muscular pathology.

Global multidisciplinary management is the essential foundation for treatment strategy. Full involvement of the surgical team from the outset helps avoid the insidious evolution toward severe deformity that occurs in absence of treatment.

Conservative management is often difficult and demanding. It may be poorly tolerated and further impair the functional potential of the disabled child or adolescent.

Surgical treatment provides a definitive and radical but effective solution in severe deformity. It is not ''pointlessly aggressive'', as physicians and physiotherapists still tend to see it, but provides real benefit, both functionally and in terms of pain. It is, however, only the last stop in a long period of preparation and assessment, weighing benefit against risks that are usually under control.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

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